Case Report

Isolated subacromial-deltoid bursitis as first manifestation of Brucellosis

Abstract

Described first by Kennedy JC in 1904, Brucella bursitis remains one of the rarest osteoarticular manifestations during this infection. Its prevalence was only 1-1.2% in wide series but seems to be clinically under-diagnosed. Indeed, its systematic screening by joint ultrasound objectified it in 5.2% of cases in some series. Shoulder bursitis (subacromial, subdeltoid or subacromial-deltoid) remains exceptional and unusual in brucellosis with only few sporadic cases in world literature. This clinical presentation of brucellosis represents a real diagnostic challenge for clinicians. We report an original observation of subacromial-deltoid bursitis as the first manifestation revealing septicemic brucellosis in 21-year-old Tunisian man.

Keywords: subacromial-deltoid bursitis, brucellosis, arthritis, shoulder, brucella

Volume 14 Issue 4 - 2022

Safa Trabelsi,[1] Souha Khfifi,[1] Rim Ben Amor,[1,2] Houcine Mrad,[1,2] Salem Bouomrani[1,2]

[1]Department of Internal medicine, Military Hospital of Gabes, Tunisia

[2]Sfax Faculty of Medicine, University of Sfax, Tunisia

Correspondence: Dr Salem Bouomrani, Department of Internal medicine, Military Hospital of Gabes, Gabes 6000, Tunisia, Email salembouomrani@yahoo.fr

Received: July 05, 2022 | **Published:** July 20, 2022

Introduction

Brucellosis, also called Malta fever or sudoro-algic fever, is an anthropozoonosis that is widely responded around the world.[1,2] It is still endemic in several countries of the Mediterranean basin, Middle East, West Asia, Africa and Latin America with an average global annual incidence estimated at 500,000 new cases per year.[1,2]. In some of these countries, the prevalence can even reach 203 cases per 100,000 inhabitants.[3] Human brucellosis is characterized by a large clinical polymorphism making its diagnosis a real challenge for clinicians, particularly those in the first line.[4,5]

Certain clinical presentations are exceptional and unexpected during this infection, particularly in the acute septicaemic forms; they are thus qualified as "unusual" by several authors.[6–8] Among these unusual manifestations of brucellosis are: uveitis, neurobrucellosis, peritonitis, orchitis, epididymitis, pericarditis, pancytopenia, myocarditis and vasculitis.[6–12] The overall frequency of these unusual clinical presentations does not exceed 5% in large series.[6–12] Bursitis remains exceptional during this infection. We report the original observation of subacromial-deltoid bursitis as the first manifestation revealing septicemic brucellosis.

Case presentation

A 21-year-old Tunisian man, with no significant pathological history, consulted for acute fever for a week with a nocturnal recrudescence with pain in the left shoulder. He was initially suspected of having covid-19 (in the middle of the second wave of the disease in our country) but the rapid diagnostic test and the specific serology were negative on several occasions. Chest CT did not show any specific signs of this infection either. Due to the persistence of the fever and the aggravation of the pain in the left shoulder with the gradual appearance of a limitation in his area of mobility, he comes to our consultation.

The questioning revealed the notion of familial brucellosis (7 members were affected) with the death of a cousin due to septicaemic brucellosis with severe hepatosplenic involvement. The patient himself reported the consumption of fresh unpasteurized milk. The examination noted a fever of 38.5°C, preserved hemodynamic, respiratory and neurological states, a painful left shoulder on mobilization, with particular limitation of abduction and external rotation and significant synovitis. No adjacent skin lesions or locoregional adenopathies were noted. Similarly, no inflammation of the other joints was noted.

Biology showed a mild inflammatory syndrome with an erythrocyte sedimentation rate of 39mmH1 and a C-reactive protein of 10mg/l. Complete blood count, uric acid, serum calcium, creatinine, muscle enzymes, blood glucose, lipid parameters, transaminases, plasma protein electrophoresis, urine sediment analysis, and thyroid hormones were within limits from normal. The radiograph of the left shoulder showed widening of the juxta-articular soft tissues without bone lesions. Ultrasound of the shoulder showed intra-articular effusion in the bicipital gutter and subacromial-deltoid bursa of moderate abundance, hypoechoic and heterogeneous and without tendon lesions. MRI confirmed subacromial-deltoid bursitis (minimal effusion in the bursa with spontaneous T2 hypersignal and T1 hyposignal) (Figures 1 & 2) with signs of minimal tendinopathy at the supraspinatus insertion.

Figure 1 MRI of the left shoulder, lateral T2-weighted view: spontaneous hypersignal of the subacromial-deltoid bursa (bursitis).

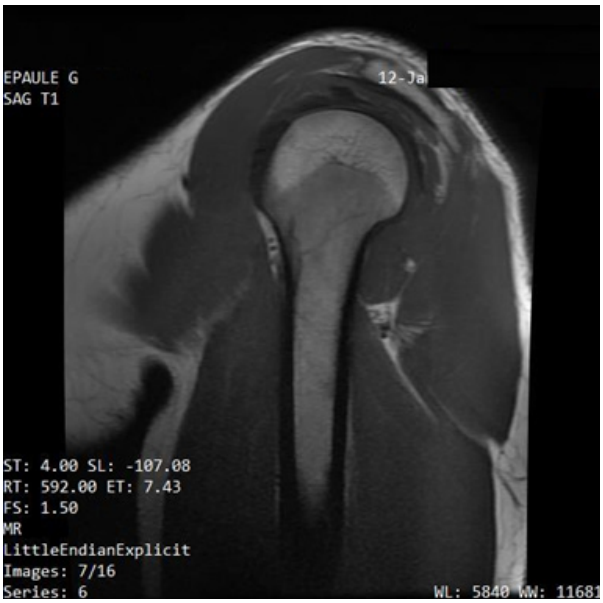

Figure 2 MRI of the left shoulder, sagittal T1-weighted view: hyposignal of the subacromial-deltoid bursa (bursitis).

The study of the puncture fluid of the subacromial-deltoid bursa revealed a fluid rich in non-specific inflammatory cells, particularly neutrophils, lymphocytes and histiocytes. Direct examination, culture, search for microcrystals and acid-alcohol-resistant bacilli were negative. Wright's serology was strongly positive at 1/5120 confirming the diagnosis of acute brucellosis. Thus the diagnosis of subacromial-deltoid bursitis revealing acute brucellosis was retained.

The patient was treated with Rifampicin (600 mg/day) and Doxycycline (200 mg/day) with a favorable outcome: apyrexia and disappearance of shoulder pain from the third day, normalization of C-reactive protein at the end of the first week and regaining full mobility of the shoulder in the second week. The follow-up ultrasound at one month showed the complete disappearance of the intra-articular effusion. No recurrence has been noted for two years now.

Discussion

Bone and joint involvement is the most frequent complication of human brucellosis.[13–15]. Its prevalence is estimated according to the series at 10-85%.[13] The most classic manifestations of osteo-articular brucellosis are: spondylodiscitis, sacroiliitis, spondylitis, discitis, septic arthritis and chronic osteomyelitis.[13–16] Abarticular manifestations are much rarer, with types of: synovitis, brusitis, tendinitis, tenosynovitis and enthesitis.[13,14,16] The most commonly affected peripheral joints are the knees, hips and ankles. The other articular sites, and particularly the shoulder, remain exceptional.[13–15,17]

Shoulder involvement in brucellosis is often described as unusual.[18] Indeed, its frequency was only 0.6% in the Iranian series by Ebrahimpour S et al, of 464 cases of brucellosis.[15] Similarly, shoulder involvement accounted for only 3.62% of brucellosis monoarthritis in the Macedonian series by Bosilkovski M et al. of 331 cases of brucellosis with mono-articular involvement.[17] Shoulder involvement may be the only clinical manifestation of acute brucellosis, making diagnosis very difficult.[19]

Brucella bursitis was first described by Kennedy JC in 1904[20] and remains one of the rarest osteoarticular manifestations during this infection.[21]. Indeed, its prevalence was only 1% in the Turkish series of Mermut G et al, of 231 patients with brucellosis[16] and 1.2% in the Kuwaiti series of Mousa AR et al, of 169 cases of brucellosis.[14] However, it seems to be clinically under-diagnosed since its systematic search by joint ultrasound had objectified it in 5.2% of the 251 patients with brucellosis in the Turkish series by Pourbagher A et al.[22]

Shoulder bursitis (subacromial, subdeltoid or subacromial-deltoid) remains exceptional in brucellosis and is often reported as sporadic cases.[21,23] Indeed, in the Turkish series by Taşova Y et al, of 238 cases of brucellosis with osteoarticular involvement, only two cases of subacromial bursitis were noted (0.84%).[24] Similarly, in the Turkish series by Pourbagher A et al, of 251 cases of brucellosis, only three had subacromial-deltoid bursitis (1.2%).[22] Finally, the systematic review of the literature carried out in 2019 by Çatal B only found six published cases of shoulder bursitis during this infection.[21] Brucella bursitis represents a real diagnostic challenge even in endemic regions, particularly the isolated and inaugural forms.[21] Its evolution is usually favorable under appropriate antibiotic therapy.[21,23]

Conclusion

As rare as it is, this unusual clinical presentation of brucellosis deserves to be known by all health professionals.

Our observation is distinguished by the isolated and revealing character of the brucella subacromial-deltoid bursitis. In endemic countries for this infection, the diagnosis of brucellosis should be discussed for any unproven bursitis.

Acknowledgments

None.

Conflicts of interest

None.

References

1. Khurana SK, Sehrawat A, Tiwari R, et al. Bovine brucellosis - a comprehensive review. *Vet Q*. 2021;41(1):61–88.
2. Shakir R. Brucellosis. *J Neurol Sci.* 2021;420:117280.
3. Wang XH, Jiang H. Global prevalence of human brucellosis. *Zhonghua Liu Xing Bing Xue Za Zhi.* 2020;41(10):1717–1722.
4. Franco MP, Mulder M, Gilman RH, et al. Human brucellosis. *Lancet Infect Dis*. 2007;7(12):775–786.
5. Ulu-Kilic A, Metan G, Alp E. Clinical presentations and diagnosis of brucellosis. *Recent Pat Antiinfect Drug Discov*. 2013;8(1):34–41.
6. Hatipoglu CA, Yetkin A, Ertem GT, et al. Unusual clinical presentations of brucellosis. *Scand J Infect Dis.* 2004;36(9):694–697.
7. Türksoy O, Tokgöz H, Toparli S. Unusual clinical presentations of brucellosis. *Scand J Infect Dis.* 2005;37(10):784.
8. Starakis I, Mazokopakis EE, Bassaris H. Unusual manifestations of brucellosis: a retrospective case series in a tertiary care Greek university hospital. *East Mediterr Health J*. 2010;16(4):365–370.
9. Gatselis NK, Makaritsis KP, Gabranis I, et al. Unusual cardiovascular complications of brucellosis presenting in two men: two case reports and a review of the literature. *J Med Case Rep.* 2011;5:22.
10. Bouomrani S, Belgacem N, Ben Hamad M, et al. Bilateral Panuveitis Revealing Acute Septicemic Brucellosis. *EC Ophthalmology.* 2018;9.6:437–441.
11. Bouomrani S, Dey M, Ahmed A. Acute Renal Failure Complicating Septicemic Brucellosis. *J Pathol Infect Dis*. 2019;2(2):1–2.

12. Bouomrani S, Mrad H, Ben Teber S. Cutaneous leuckocytoclastic vasculitis revealing acute brucellosis. *American Journal of Medical Case Reports*. 2021;6(9):335–338.

13. Esmaeilnejad-Ganji SM, Esmaeilnejad-Ganji SMR. Osteoarticular manifestations of human brucellosis: A review. *World J Orthop.* 2019;10(2):54–62.

14. Mousa AR, Muhtaseb SA, Almudallal DS, et al. Osteoarticular complications of brucellosis: a study of 169 cases. *Rev Infect Dis*. 1987;9:531–543.

15. Ebrahimpour S, Bayani M, Moulana Z, et al. Skeletal complications of brucellosis: A study of 464 cases in Babol, Iran. *Caspian J Intern Med.* 2017;8(1):44–48.

16. Mermut G, Ozgenç O, Avcı M, et al. Clinical, diagnostic and therapeutic approaches to complications of brucellosis: an experience of 12 years. *Med Princ Pract*. 2012;21(1):46–50.

17. Bosilkovski M, Zezoski M, Siskova D, et al. Clinical characteristics of human brucellosis in patients with various monoarticular involvements. *Clin Rheumatol.* 2016;35(10):2579–2584.

18. Oteiza J, Barasategui JI, Torroba L, et al. Unusual localization of brucellosis related arthritis of the shoulder. *Rev Clin Esp.* 2001;201(11):678–379.

19. Sánchez Rodríguez A, Sánchez Fuentes D, Pedraz Calvo M, et al. Osteoartritis supurada de la articulación del hombro como única manifestatión de una brucelosis aguda [Suppurating osteoarthritis of the shoulder joint as the only manifestation of acute brucellosis]. *Rev Clin Esp.* 1982;166(3-4):165-7.

20. Kennedy JC. Notes on a case of chronic synovitis, or bursitis, due to organism of the Mediterranean fever. *J R Army Med Corps* 1904;2:178–180.

21. Çatal B. Rare form of brucellosis, subacromial and subdeltoid bursitis: A case report and literature review. *Eklem Hastalik Cerrahisi.* 2019;30(3):333–337.

22. Pourbagher A, Pourbagher MA, Savas L, et al. Epidemiologic, clinical, and imaging findings in brucellosis patients with osteoarticular involvement. *AJR Am J Roentgenol.* 2006;187(4):873–880.

23. Wang FS, Shahzad K, Zhang WG, et al. Atypical presentation of shoulder brucellosis misdiagnosed as subacromial bursitis: A case report. *World J Clin Cases*. 2021;9(4):927–934.

24. Taşova Y, Saltoğlu N, Sahin G, et al. Osteoarthricular involvement of brucellosis in Turkey. *Clin Rheumatol. 1999*;18(3):214–219.

Citation: Trabelsi S, Khfifi S, Ben Amor R, et al. Isolated subacromial-deltoid bursitis as first manifestation of Brucellosis. *MOJ Orthop Rheumatol.* 2022;14(4):97–99. DOI: 10.15406/mojor.2022.14.00586